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Advanced osteitis fibrosa cystica in the absence of phalangeal subperiosteal resorption

A case report and review of the literature

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Summary

A case of primary hyperparathyroidism with advanced osteitis fibrosa cystica but without any subperiosteal phalangeal bone resorption is described. A review of this unusual radiological feature is presented. High-detail magnification radiography (microradiography) is advocated for the early diagnosis of bony defects in hyperparathyroidism.

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Osteitis fibrosa cystica refers to the advanced bony changes seen in hyperparathyroidism. It is characterized radiologically by the presence of subperiosteal resorption of cortical bone, grossly abnormal bone architecture, bone cysts and, in severe cases, the development of brown tumours.¹ Brown tumours contain localized accumulations of giant cells and fibrous tissue. A common biochemical accompaniment of this condition is a raised plasma alkaline phosphatase level of bone origin.² The initial effect of parathyroid hormone excess on bone is an increase in bone resorption caused either by osteocytic osteolysis or by osteoclastic surface resorption.³ The former cannot

be identified by fine-detail radiography, but the earliest manifestation of osteoclastic surface resorption can be seen radiographically as subperiosteal resorption of the cortices of the radial aspects of the middle phalanges of the hands and as intracortical resorption (also known as 'haversian resorption' or 'cutting cones').³

Radiographic evidence of osteitis fibrosa cystica in the shoulder, pelvic girdle, axial skeleton or skull represents more advanced disease than does phalangeal subperiosteal resorption⁴ and therefore brown tumours are rarely encountered in the absence of phalangeal subperiosteal resorption in the hands.^{1,5} This article describes such an entity in a patient diagnosed as having primary hyperparathyroidism.

Case report

A 51-year-old woman had a 6-month history of malaise and recent onset of left renal colic. An excretory urogram demonstrated a ureteric calculus, which was removed and found to be composed predominantly of calcium. Biochemical investigations revealed a raised total serum calcium level of 2,8 mmol/l (normal 0,1 - 1,4 mmol/l), a low to normal serum phosphate level of 0,79 mmol/l (normal 0,8 - 1,4 mmol/l), and a persistently raised total serum alkaline phosphatase level of 120-130 IU/l (normal 30 - 100 IU/l). The level of plasma parathyroid hormone measured by C-terminal radio-immunoassay was significantly increased and ranged from 290 to 320 pmol/l (normal 0 - 130 pmol/l). Renal function was normal.

A radiographic skeletal survey showed vertebral biconcavity and increased vertical striations, indicating axial osteopenia. The mandible demonstrated multiple medullary cystic lesions with thinning of the cortex (Fig. 1). A fine-detail radiograph of the hands was examined with a 6-power magnifier with a built-in 0,1 mm scale in order to measure cortical widths and to detect cortical erosions.

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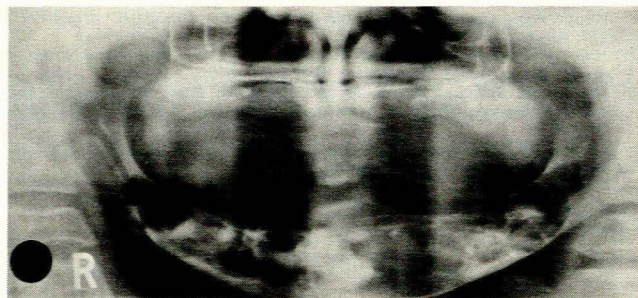


Fig. 1. Orthopantomogram showing multiple cysts in the mandible with thinning but not destruction of the cortex. A biopsy specimen from the cysts showed histological features in keeping with brown tumours.

The Exton-Smith index of metacarpal cortical area⁴ was markedly below the 10th percentile for the patient's age and sex, indicating severe osteopenia. No subperiosteal phalangeal bone resorption could be seen (Fig. 2). The rest of the skeleton was radiologically normal. On histological examination the cystic lesions in the jaw showed localized collections of fibrous tissue and giant cells in keeping with the pathological diagnosis of brown tumours. Four hyperplastic parathyroid glands were removed from the patient's neck.

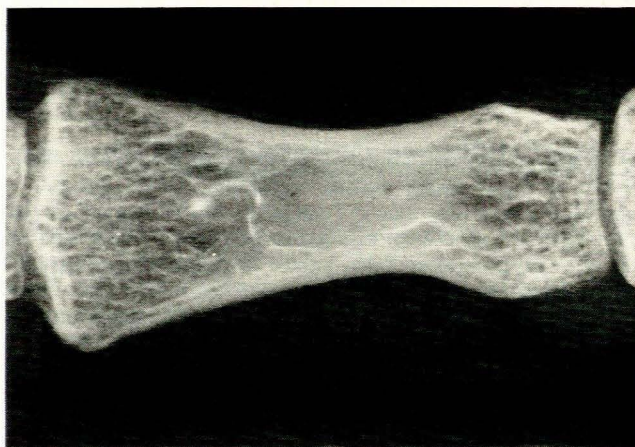


Fig. 2. High-detail microradiograph of a middle phalanx of the right hand showing no subperiosteal resorption or increased intracortical cutting cones.

Discussion

High-detail magnification radiography (microradiography) of the hands is essential in the early diagnosis of bony defects in hyperparathyroidism. It has a diagnostic accuracy of 50 - 60%, in comparison with 0 - 29% for ordinary routine hand radiographs in different series.^{6,7} This method can be refined even further by quantitating the subperiosteal resorption since very early cases are difficult to distinguish from the periosteal irregularity in the metaphyses of normal phalanges.³

Osteopenia as part of hyperparathyroid skeletal involvement, or as the presenting feature of this bone disease has often been described.^{1,8} The Exton-Smith index of cortical area is a valid means of assessing bone mineral content.⁴ By using this index on the second metacarpal of the hand, abnormal cortical thinning could be detected in 83 - 86% of 162 patients with primary hyperparathyroidism.² In this context it is possible that osteopenia is an earlier, albeit nonspecific, sign of hyperparathyroid bone disease than subperiosteal resorption.

Increased intracortical resorption is a nonspecific sign of diseases with high bone turnover, and is found in hyperparathyroidism, hyperthyroidism and acromegaly,⁹ among others. In our patient intracortical cutting cones ('haversian resorption') were accurately quantitated as described in the literature³ but were found to be within normal limits, despite the presence of a raised serum alkaline phosphatase level.

In hyperparathyroidism brown tumours, i.e. localized areas of intense osteoclastic activity, are rarely encountered in the absence of phalangeal subperiosteal resorption.^{1,10} Pugh⁵ described 2 such cases of primary hyperparathyroidism in which radiological evidence of brown tumours was present without evidence of subperiosteal phalangeal resorption. No high-detail magnification radiography was done in these cases and the very early signs of hyperparathyroid bone disease (grade I or II subperiosteal resorption of the middle phalanges)³ could therefore have been missed.

Finally, it is postulated that severe hyperparathyroidism leads to osteitis fibrosa cystica due to increased osteoclastic activity and that the milder form of the disease results in osteocytic osteolysis with osteopenia and thinning of the cortices.^{1,10}

This case of primary hyperparathyroidism was therefore unusual in that the patient had brown tumours, a sign of advanced osteitis fibrosa cystica, without any evidence of phalangeal subperiosteal resorption, a well-accepted early feature of hyperparathyroid bone disease. We further stress the importance of microradiography in the early radiological detection of such bone disease.

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